Isolated Fallopian Tube Torsion: A Rare Twist with a Diagnostic Challenge That May Compromise Fertility

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Isolated Fallopian tube torsion (IFTT) is a rare entity with utmost important gynecological emergency with difficult preoperative diagnosis. Our aim is to analyze the clinical presentation, risk factors, and management of IFTT. We retrospectively analyzed all patients with intraoperative diagnosis of IFTT in our endogynecological department over a time period of 3 years and 6 months (January 2015-June 2018) in a tertiary level laparoscopic center. The clinical profile of the patients was analyzed and the results formulated. Statistical analysis was done by SPSS system, Version 15.00 (SPSS Inc., Chicago). A total 17 cases were diagnosed with IFTT with or without pathology. The mean age was 28.07 ± 11.3 years. Lower abdominal pain was the most common symptom (88%). About 47% had a history of tubal ligation. Salpingectomy was done in the majority of the patients (82.3%). Detorsion and preservation of the tube was possible in 17.6% of the cases. High index of suspicion is needed to diagnose this rare cause of acute abdomen. Hence, an early intervention can enhance the salvageability of the affected tubes which has a positive impact on the fertility status of the patients. Tubal preservation is the preferred procedure of choice whenever feasible.

KEYWORDS: Isolated Fallopian tube torsion, laparoscopy, salvageability

Introduction

solated Fallopian tube torsion (IFTT) is a most Limportant surgical emergency in young adolescents and reproductive age group with a reported incidence of 1/1.5 million.[1] It may occur due to intrinsic factors such as congenital anomalies, long or spiral tube, hydrosalpinx, pelvic inflammatory disease (PID), tubal ligation or extrinsic factors such as adhesions, adnexal venous congestion, gravid uterus, uterine, ovarian or para ovarian masses, and trauma.^[2] Laparoscopy is the gold standard for accurate diagnosis and management. However, the diagnosis is rarely made and surgery often delayed due to extremely difficult preoperative diagnosis because of varied clinical presentation and nonspecific imaging findings. Early diagnosis and immediate surgical intervention are crucial in these patients to avoid salpingectomy.

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The purpose of this study is to describe the clinical presentation, triggering factors, diagnostic tests used, and the management of surgically proven IFTT.

MATERIALS AND METHODS

We have carried out this retrospective study in our tertiary level laparoscopic center over a period of 3 years and 6 months (January 2015–June 2018) after obtaining informed written consent from all the patients/parents. This study was performed after our ethical committee approval. Patients who were confirmed to be an IFTT intraoperatively irrespective of their age group were included in this study. Ethical approval for this

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study (GEM/REC/2018) was provided by the Research Ethics Committee of GEM Hospitals and Research institute on December 03/2018. "All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards."

There were 17 cohorts with IFTT diagnosed intraoperatively. All the clinical parameters such as demographic details, previous history of pelvic surgery, previous adnexal torsion, triggering factors, duration of symptoms, ultrasonography (USG) findings, tumor markers, time from onset of symptoms to surgery, and admission to surgery interval were recorded, analyzed, and formulated. All patients had transvaginal ultrasound except in unmarried women. All patients underwent laparoscopy. Data regarding intraoperative findings such as site, side of torsion, grading of ischemia or necrosis, and number of twists were noted. Grade of ischemia and viability was assessed after detorsion.

Grading of ischemia was done based on the Parelkar grading

- Grade 1: Slightly discolored, normal sized which promptly reverted to normal color after detorsion
- Grade 2: Dark red to brown in color, mildly enlarged, and edematous which became hyperemic after detorsion
- Grade 3: Brown to black in color, grossly enlarged, and edematous with a slight improvement in color after detorsion
- Grade 4: Completely black, necrotic, and no improvement in color after detorsion.^[3]

To reduce the subjective operator bias, surgeries were performed by two laparoscopic gynecologists. Detorsion was attempted in all the patients who have not completed family and patients who have completed their family underwent salpingectomy irrespective of the grade of ischemia.

Our primary outcome of this study was to describe the clinical presentation, triggering factors, and diagnostic tests findings in women with surgically proven IFTT.

Our secondary outcome was to assess the feasibility of tubal preservation in those who were desirous to conceive.

Following the surgery, all patients were clinically assessed for pain, need for analgesics in the first 24–48 h.

To reduce further compromise of fertility, these patients were followed up with ultrasound and early warning signs of torsion were explained to all patients.

Ultrasound with Doppler was done to look for recurrent torsion of ipsilateral or contralateral adnexa during 1-month, 6-month, and 1-year postoperative follow-up. Tubal patency was assessed with hysterosalpingography (HSG) in married women following 1 month of detorsion. All the data were recorded in an Excel sheet, and the required variables were expressed in mean, range, and standard deviation.

RESULTS

The study group included 17 patients with surgically confirmed IFTT. Characteristics of the patient and triggering factors for IFTT are shown in Table 1. The age of the patients ranged from 11 to 46 years. The mean age was 28.07 ± 11.3 years. A total number of unmarried patients were nine, of which two patients had a history of hydrosalpinx, one patient had a history of PID, and none had a history of previous pelvic surgeries. All married women (n = 8) had a history of prior pelvic surgeries such as Lower Segment Cesarean Section (LSCS) and tubal ligation. In our study group, two were premenarcheal and one had primary amenorrhea, and in the remaining 14 patients, nine were in the premenstrual period, three were in the menstruating phase, and two were in the day 8 of the cycle at the time of presentation.

Signs and symptoms are shown in Table 2. On clinical examination, 14 patients had lower quadrant tenderness and five of them had tender palpable mass on vaginal/rectal examination. The duration of symptoms ranged from 1-day to 30 days. The mean time from initial examination to surgical intervention ranged from 1 h to 24 h.

Ultrasound was done transvaginally in all married women. Huge adnexal cysts or hydrosalpinx of ≥7 cm were seen in 10 patients, of which four were hydrosalpinx and the remaining six were ovarian/ paraovarian cysts. Tumor markers were done in those six patients, which were reported to be normal. Ultrasound was showing free fluid in the pelvis with hydrosalpinx in one patient. The presence of blood flow in adnexa was assessed by color Doppler in 14 patients and all of them had normal flow, and the whirlpool sign was not assessed in any of our patients. Preoperative diagnosis of either adnexal or Fallopian tube torsion was not made by imaging modalities. Clinically, only three were diagnosed as Fallopian tube torsion preoperatively and 11 were misdiagnosed as adnexal torsion [Table 3]. In the remaining three patients, preoperative diagnosis was hydrosalpinx/ovarian cysts and all were ≥35 years, of which two were asymptomatic.

Intraoperative findings are shown in Table 4. Intraoperatively, free fluid in the pelvis was noted in

Table 1: Characteristics of 17 patients with isolated Fallopian tube torsion

Characteristics	Number of patients (n=17
Age distribution (in years)	
11-19	6
20-40	9
41-46	2
Menopause	0
Desirous of fertility	9
Not desirous of fertility	8
(multiparous- completed family)	
Triggering factors	
Previous LSCS with sterilization	6
Prior tubal ligation	2
Posthysterectomy	0
No previous pelvic surgeries	9
Previous history of PID	3
Previous history of hydrosalphinx	4
History of ovarian cyst	0

PID=Pelvic inflammatory disease, LSCS=Lower segment cesarean section

Table 2: Clinical presentation		
Signs and symptoms	n = Number of patients	
Nausea	2	
Vomiting	6	
Dysuria	3	
Fever	4	
Abdominal pain	15	
Diffuse	7	
Localized	8	
Sharp sudden severe pain	9	
Recurrent pain	3	
Right lower quadrant pain	5	
Left lower quadrant pain	4	
Periumbilical pain	1	
Back pain	2	
Asymptomatic	2	
Signs		
Abdominal tenderness	14	
Palpable mass	5	

eight patients, of which five were hemorrhagic. All patients had normal ovaries except two patients who had simple ovarian cyst.

Salpingectomy was done in 14 of 17 patients (82.3%). Detorsion was attempted in all unmarried nine girls, and we were able to preserve three tubes with Grade 1 and 2 ischemia. The remaining six patients had irreversible ischemia following detorsion. Hence, salpingectomy was done in these patients.

Histopathologically, all were benign hydrosalpinx/paratubal cysts. In our study, a 25-year-old unmarried girl presented with sudden onset of right lower abdominal

pain and tenderness with nausea and vomiting; as her ultrasound did not show any abnormality, she underwent contrast-enhanced computed tomography which also failed to show adnexal torsion. With clinical suspicion of adnexal torsion, diagnostic laparoscopy was done which showed normal tube undergoing torsion with Grade 2 ischemia with chronically inflamed appendix in retrocecal position adherent to cecum which could be the triggering factor for isolated right Fallopian tube torsion. The tube was preserved following detorsion in this patient.

None of the patients had a recurrence in our study during the postoperative follow-up. The three patients for whom tubes were preserved had completed their 1-year follow-up and had no recurrence. Among the three patients, tubal patency was assessed by HSG in two patients after their marriage and bilateral tubes were patent in both the patients and the third patient is on regular follow-up. For her, HSG was planned after her marriage.

DISCUSSION

The diagnosis of Fallopian tube torsion is very often missed due to the lack of pathognomonic features; hence, it is a diagnostic challenge for the clinician to differentiate this from ovarian torsion, PID, ectopic pregnancy, appendicitis, degenerating myoma, or urinary condition. In our study, lower abdominal pain was the predominant symptom in 88% of the patients (15 out of 17), similar to Demirel et al.[4] Most of them had delayed diagnosis as a result of the atypical presentation. About 47% (8/17) of our study population had nausea and vomiting, which could be due to visceral response to torsion, which further confuses this entity with acute appendicitis. Few patients had dysuria in 17.6% (3/17) and diarrhea in 11.7% (2/17) of the patients, which may represent irritation from the hemorrhagic and necrotic process. Laboratory findings were nonspecific. Leukocytosis was present in 41.1% (7/17) of our patients and fever was rarely present. Necrosis can cause fever, leukocytosis, elevated erythrocyte sedimentation rate, and C-reactive protein. The classical finding of Fallopian tube torsion is a sudden onset of sharp severe pain in the abdomen radiating to the flanks which was seen in 52.9% (9/17) of the patients.

Tubal ligation has been reported as one of the predisposing factors for IFTT. Following tubal ligation secretions retained in the distal end may lead to formation of hydrosalpinx. This post surgical adhesion will lead to formation of a non mobile nidus. This combination, might predispose to torsion of tube around the nidus, although the exact pathogenesis is unknown. Hence, whenever we encounter an adnexal torsion, IFTT should be suspected in patients with a previous history

Table 3: Pre-operative workup and diagnosis		
Pre operative work up	n = Number of patients	
Leukocytosis (≥10,000)	7	
USG findings		
Ovarian cyst	7	
Hydrosalphinx	6	
Adnexal cyst	2	
Para-ovarian cyst	1	
Normal study	1	
Doppler done	14	
CT scan done	7 (diagnosis not conformed in all 7)	
MRI done	1 (diagnosis not confirmative)	
Free fluid	8 (intra-operative)	
	1 (USG)	
Mean duration of	4.9±9.5	
symptoms (days)		
Symptoms onset to surgery	13.94	
(days)		
Admission to surgery interval (min)	624.7 min (10.4 h)	
Preoperatively diagnosed as		
Fallopian tube torsion-3	Intraoperatively, all 17 were	
Ovarian torsion-3	Fallopian tube torsion	
Adnexal torsion-8		
As hydrosalpinx-1		
As ovarian cyst-2		

USG=Ultrasonography, MRI=Magnetic resonance imaging, CT=Computed tomography

of tubal ligation, with an elongated cystic mass, who have a sudden onset of abdominal pain or pelvic pain. About 47% of the patients (8/17) in our study who were married and completed family had a history of pelvic surgery with tubal ligation.

In adults, triggering factors for hydrosalpinx and IFTT are PID, previous abdominal surgeries, history of tuberculosis, previous ectopic pregnancies, and endometriosis. Hydrosalpinx is rarely seen in adolescents. In our study, hydrosalpinx was seen in 70.5% (12/17 patients), in which five were sexually inactive, of which two of them were premenarcheal and none of these five patients had previous abdominal surgeries. A possible etiology could be due to a congenital malformation of the tube-like long and mobile tubes, with or without an asymptomatic distal tubal occlusion, which is revealed as hydrosalpinx following activation of the reproductive axis.^[5]

In our series, we could observe IFTT appeared more frequently in the premenstrual period in 52.9% (9/17). These findings suggest that immediate premenstrual congestion of the mesosalphingeal veins would be one of the triggering factors.^[6]

Associated adnexal cysts could be the lead factor for IFTT as stated by Bertozzi et al. who described

Table 4: intra operative findings and outcomes	
Surgical findings	n = Number of patients
Side of torsion	
Right side	6
Left side	11
Number of twists	
Partial	3
Once	2
Twice	4
Three times	5
Four times	3
Mean cyst diameter (in cm)	6.8 ± 2.5
Grade of ischemia	
Grade 1	5
Grade 2	3
Grade 3	2
Grade 4	7
Associated pathology	
Hydrosalphinx	12
Paratubal cyst	4
Torsion of a normal tube	1
Detorsion attempted	9
Tube salvaged	3 (2 paratubal cyst, 1 normal tube)
Surgery done	
Salphingectomy	14
Salphingostomy	Nil
Detorsion±paratubal	3
cystectomy	
Histopathology	
Hydrosalphinx	12
Paratubal cyst	4
Nil	1

21 cases of hydrosalpinx in their study and is the most common cause for Fallopian tube torsion, similar to our study showing 70.5% (12/17). Paratubal cyst in 23.5% (4/17) was the second common cause for tubal torsion in our case series. Paratubal cysts or hydatids of Morgagni which are remnants of Mullerian or Wolffian structures have been described as a cause for Fallopian tube torsion described by Mueller and Tomita. Mechanical dragging of the Fallopian tube and an increase in the heaviness of the distal end of the fimbria due to the cyst can be a possible explanation.

Adnexal torsion is more common on the right side due to the preventive effect of the sigmoid colon on the left side acting as a cushion, dextrorotation of the uterus, and relatively increased venous flow on the left causing less congestion. Another reason may be due to more frequent exploration of right-sided pathology with a suspicion of appendicitis. In our series, IFTT however occurred on the left side in 64.7% (11/17), similar to the two largest case series reported, which may suggest that IFTT occurs more commonly on the

left side,^[5,7] though larger case series are needed for confirmation.

Ultrasound should be the first imaging modality of choice due to the lack of radiation and it is useful to discriminate other causes of acute abdominal pain, but unfortunately, accurate diagnosis of IFTT is possible in only 30% of the cases and the torsed tube commonly mimics like a multicystic adnexal or ovarian cyst.[9] The value of CT and MRI in diagnosing IFTT is doubtful and controversial with very little benefits and may delay the immediate surgical intervention. If USG is indeterminate, Contrast-enhanced MRI may be useful, to identify (1) the twisted vascular pedicle, (2) uterine deviation to the twisted side (3) thickened and enhanced Fallopian tube, (4) also particularly useful in young adolescents or pregnant women without ionizing radiation.[10] Color Doppler may be useful in diagnosing the IFTT, but normal flow definitely will not rule out the torsion. A whirlpool sign is known for its high sensitivity in diagnosing the IFTT,[9,11] which was not assessed in our study; hence, the diagnosis could be missed and none of the patients were identified as adnexal torsion by imaging modality in our series.

Laparoscopy is the only gold standard for accurate diagnosis and treatment of IFTT.^[12] Recovery after laparoscopy is faster and also causes fewer pelvic adhesions which is particularly important for women who wish to preserve the fertility.

Laparoscopic detorsion of the tube is the treatment of choice whenever feasible to preserve the fertility. Although one Fallopian tube is sufficient for reproductive function, salpingectomy may have an adverse impact on fertility as the remaining tube may get affected by the infection, ectopic, or torsion later. Complete or partial salpingectomy is indicated if the tube is irreversibly ischemic or any suspected neoplasm or in adult patients with infertility with unilateral hydrosalpinx, salpingectomy of the affected side resulted in increased *in vitro* fertilization pregnancy rates.

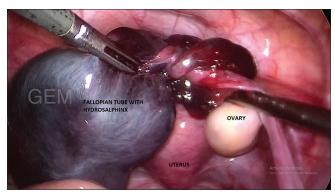


Figure 1: Right fallopian tube torsion Grade 4 necrotic tube

Concerning salpingectomy in our series, most of the torsions were noted in the distal two-third of the tube, and we believe that the complete salpingectomy is better than partial whenever it is necessary because the proximal nonfunctional part of the tube can be a site of ectopic pregnancy or reformation of hydrosalpinx if it gets occluded and removal of only ampulla and fimbria and keeping the nonfunctional portion is of no use and may increase the risk of recurrence.^[7,13] Hence, we did complete salpingectomy in all our patients [Figure 1 right Fallopian tube torsion Grade 4 necrotic tube].

The role of neosalpingostomy, especially in tubal disease like hydrosalpinx, is controversial at present and has been reported in the pediatric population, as this conservative surgery has few major concerns such as increased rate of ectopic and chance of recurrence, and also, it does not have a clear data about the functional status of the preserved tube, and future reproductive potential cannot be assessed.^[5,14] Hence, we have not tried in any of our cases.

In our study, detorsion was attempted in all the nine unmarried girls and were able to preserve the tubes in three patients with Grades 1 and 2 ischemia, in which one patient was a 12-year-old with Turner syndrome with hypoplastic uterus and streak ovaries who had paratubal cyst [Figure 2 Grade 1 ischemia with paratubal cyst]. Hence, the salvageability of the tubes in our study was 17.6%, similar to other authors such as Gaied *et al.*^[15] who had a tubal preservation rate of 12% of the cases and Bertozzi *et al.*^[7] preserved 33.3% of the tubes (7/21).

The role of salpingopexy to prevent recurrent tubal torsion is still unclear, as there is no literature to support this procedure to be safe, and it may have a negative impact on tubal function and fertility.

One limitation of this study was retrospective manner used to classify the cases, and we have included only

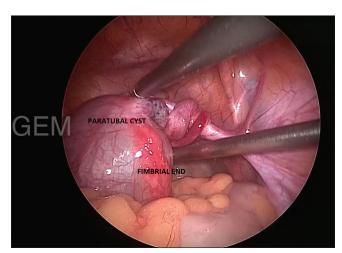


Figure 2: Grade 1 ischemia with paratubal cyst

surgically proven cases and may have missed patients with atypical presentation of IFTT who did not undergo surgery. To our knowledge, this study would be one of the largest case series, and our data would be helpful for the prompt diagnosis and treatment of this rare condition with the sparse data.

CONCLUSION

IFTT is a rare clinical entity, and diagnosis is too often missed due to varied atypical presentation despite the advanced imaging techniques. A high index of suspicion and early surgical intervention will help in salvaging the tube, which has positive impact on the fertility status. Laparoscopy remains the gold standard for the definitive diagnosis and treatment. Tubal preservation should be favored whenever possible.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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